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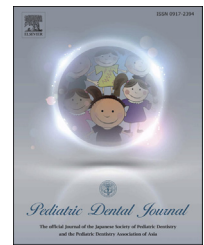
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Case Report

Ameloblastic fibro-odontoma located in the maxilla of a 3-year-old girl

Q2 **Ismail I. Ülgür^a, Rosmarie Caduff^b, Juliane Erb^c, Hubertus van Waas^c,
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ABSTRACT

Ameloblastic fibro-odontoma (AFO) is a rare benign tumor which contributes to 1–3% of all odontogenic tumors. AFO is mainly localized in the posterior region of the mandible and typical patients are children who present to their dentists due to non-erupting teeth. We describe an unusual example of a large AFO located in the upper right maxilla and maxillary sinus of a 3-year-old girl. The aims of this case report are to strengthen the knowledge of and to stimulate the discussion about AFO and to point out, that a conservative surgical procedure with regular follow up is highly recommended.

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1. Introduction

Contributing to only 1–3% of all odontogenic tumors [1–4] the ameloblastic fibro-odontoma (AFO) was first described by Hooker in 1967 and distinguished from the more aggressive ameloblastic odontoma (AO), also called ontoameloblastoma [5]. According to the revised World Health Organization (WHO) Classification of odontogenic tumors of 2005, the AFO is described as a rare mixed odontogenic tumor which is usually benign [6]. The AFO shares similarities with the ameloblastic fibroma (AF) and the ameloblastic fibro-dentinoma (AFD) but may be distinguished from these two entities by the

presence of dentin and enamel on histological examination [6–8].

A current study describes a mean age of 9.6 years which underlines the fact that AFOs are predominantly found in children, and comprise around 7% of all odontogenic tumors under the age of 16 [3,9]. Commonly, these lesions are found on routine radiography of patients with delayed eruption of teeth or in patients presenting with asymptomatic intraoral swellings [10].

Radiographs generally show well-defined radiolucent areas containing various amounts of radio opaque material of irregular size and form with the radiopaque areas dominating in most cases [4,5,10–12]. Earlier tumors may initially present

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as radiolucent areas only and could show calcification in later stages of development [13].

Even though there seems to be a clear definition after the WHO Classification of 2005 concerning AF, AFD and AFO, there are still many controversies regarding the etiology, histology, therapy and prospective outlook [4,14]. Our case report aims to further the discussion and classification of such tumors by presenting a patient with a large ameloblastic fibro-odontoma involving an unusual location, namely the maxillary sinus.

2. Case report

A three-year-old girl was referred to the Department of Pediatric Dentistry at the University of Zurich by her private dentist after the mother noted a swelling in the upper right maxilla and non-eruption of a tooth in the related area. Non-eruption of the first deciduous molar as well as a hard, tumorous entity in the described region was clinically seen by her private dentist and a large opacity was seen on the dental radiograph (Fig. 1). The dentist was concerned for an infection of the sinus cavity and referred the patient to the Pediatric Dentistry Department, University of Zurich.

The child was a healthy, well-nourished girl with an unremarkable social, medical and family history. Additionally there were no related histories or clinical signs of trauma or infection in the described area and oral inspection showed adequate oral hygiene.

Clinically there was a slight swelling in the region of teeth 54 and 55, with tooth 54 being absent. Palpatory examination however revealed a non-tender, bony hard high buccal swelling in the region of the right maxilla. The orthopantomograph revealed an erupted tooth 55 but instead of tooth 54 a complex of predominant opacities and more peripheral radiolucent areas could be detected. The lesion extended into the maxillary sinus with conforming tooth displacement (Fig. 2).

The differential diagnosis from the pediatric dentist included an odontoma and therefore the patient was sent to the Department of Cranio-maxillofacial and Oral Surgery of the University Hospital of Zurich for further examination.



Fig. 1 – Pre-operative dental film showing absent tooth 54, central radio-opaque area surrounded by a radiolucent area.



Fig. 2 – Orthopantomogram view showing the comparative obliteration of the right maxillary sinus.

Volume tomography (Fig. 3a, b and c) revealed a large calcifying tumor of the upper right maxilla involving the 54/55 area, reaching into the upper right maxillary sinus and filling out the latter nearly completely with conforming tooth displacement. A biopsy was performed under general anesthesia including bony-like fragments and firm white, gray tissue- fragments measuring $18 \times 18 \times 9$ mm.

Histopathological examination revealed a composite tumor of well-formed dentin and enamel lying in a cell rich myxoid stroma without mitotic activity consistent with the diagnosis of an ameloblastic fibro-odontoma (Fig. 4a, b).

The tumor was resected two months later under general anesthesia at the Children's Hospital of Zurich. Access to the tumor was obtained via an extended lateral buccal vestibular incision. Wide, sub periosteal exposure of the lateral and anterior wall of the maxillary sinus was achieved and the inferior orbital nerve was isolated and protected. The lateral wall of the maxillary sinus was removed and the tumor was identified from the maxillary alveolus up to the cranial extension thereof. Due to its large size, the tumor had to be segmented with a bur and chisel and then removed. Due to the clinical and radiological examination it can be presumed that the permanent tooth follicles were involved in the pathological process and partial anodontia will result in this area. The defect was not filled with any material and direct closure was achieved with resorbable sutures, avoiding the risk of an oro-antral communication.

The postoperative histological examination was identical to the biopsy done before and confirmed the diagnosis of AFO. Immediate and late post-operative healing was uneventful. Two months after the tumor ablation an additional low radiation dose volume tomogram was done, confirming complete removal of the tumor (Fig. 5a, b). In a follow up control by means of a low resolution volume tomogram six months later no recurrence was noted.

3. Discussion

This case study shows an unusual large and less common location of an AFO occurring in a three-year-old girl. In the literature only few studies are dealing with ameloblastic fibro-odontoma involving the maxillary sinus [13,15,16]. Buchner [9]

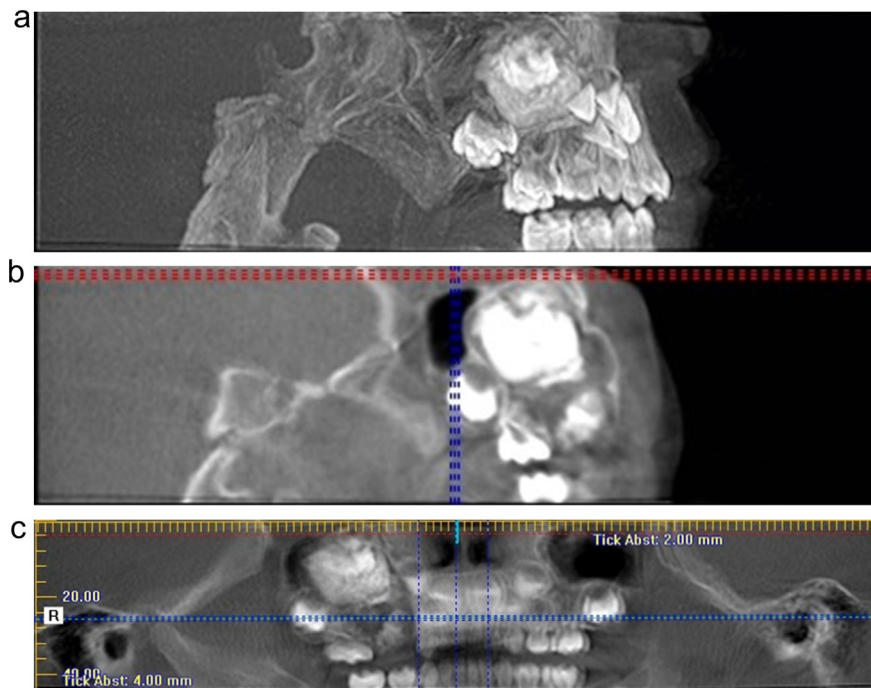


Fig. 3 – a: Sagittal volume tomogram view, demonstrating the vertical extension and tooth displacement. b: Sagittal view of the large tumor, encroaching on the maxillary sinus. c: Orthopantomogram view of the volume tomogram.

stated in a current study that the main localization of AFO was the posterior molar mandible region whereas older studies such as those of Slootweg [1] or Shafer [17] suggested no difference in the frequency between maxilla and mandible. The reason for these conflicting findings may be the fact that in former studies [1,3,4,17] the number of papers published on this topic was rather low. In the meta-analysis of Boxberger et al., reviewing 208 cases there was an incidence of 25.2% in the posterior maxilla. Nowadays, more and more reports favor the mandible as the more frequent anatomical region [13].

AFO occurs especially in children and young adults with a mean age of 8–11 years with a slight male preponderance [1,9,13]. Our patient is a very young girl presenting to our department at age of three with an extensive tumor. Similar descriptions in the literature of young patients by other authors such as Dhanuthai [18] of a one year old child in Bangkok, Thailand with an AFO are found, but are very rare.

Comparing our case with the literature [11,19–22] we describe one of the youngest patients to now. Interestingly, patients coming from countries such as India [19,20] or South America [11,21,22] present to dentists often at teenage or adult age whereas in countries such as Germany [23,24] or Switzerland, tumors are detected much earlier. An important explanatory reason may be the fact that the latter countries have well developed health care systems and dental examinations are offered in early childhood. Our patient was first examined by her private dentist at the age of two due to a non-erupting tooth.

There is no consensus in the literature on the discussion whether AF, AFD and AFO are separate entities or are different stages in the maturation of the same entity [4]. According to Cahn and Blum AF, AFD and AFO represent various stages of the same lesion and therefore will mature with time and ultimately develop into an odontoma [25]. On the other hand,

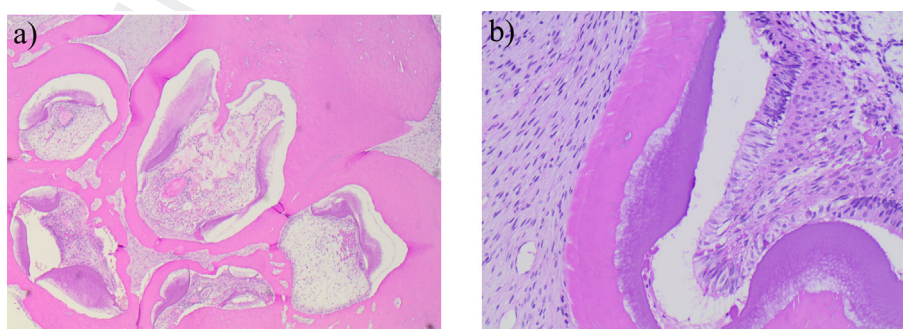


Fig. 4 – a: Overview (H&E). Well-formed dentin and enamel. b: Dentin and enamel formation (right side), cell rich loose stroma (left side) (H&E).

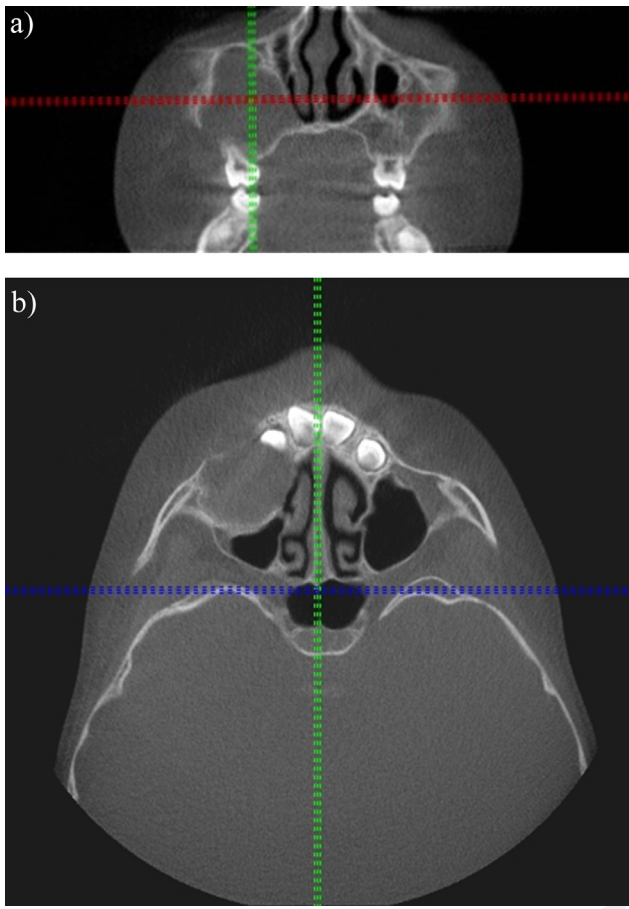


Fig. 5 – a and b: Post-operative coronal and axial views demonstrating complete removal of the tumor.

many studies available support the classification of the WHO 2005 presenting the AFO as a separate entity without progression to an odontoma [6].

This view is supported by a review study [26] which demonstrates that 92.7% of all reported recurrent AFs showed no evidence of maturation into a more differentiated odontogenic lesion such as AFO. In general, a tendency towards acceptance of the WHO histological classification can be observed, as in the case of our case presentation as well. In accordance with the WHO classification our diagnosis of AFO was supported by the existence of enamel and dentin in the histopathological examination.

This case study supports the attitude of conservative and non-aggressive approach as described in the literature because AFO are non-malignant lesions and the probability of recurrence can be described as low. As reported above a small part of the maxillary sinus was exposed and partially involved in the surgical resection. A more aggressive treatment as described by Gिरradi concerning fibrodentinoma [20] is not recommended because of increased morbidity. Especially in the mandible, the risk of a pathological fracture is very high when performing a marginal resection and thus cannot be supported when compared to a more conservative procedure of tumor tissue removal [14]. At this age involvement of primary teeth is the rule rather than an exception. Displacement

or even absence of permanent teeth as in this case may occur and needs careful follow up.

Friedrich [27] described a malignant and aggressive transformation 18 months after treatment and Furst [28] presented a case with a recurrence two years after surgery. An average recurrence rate of 7.4% is reported [13]. Howell and Burks demonstrated 2 cases of ameloblastic fibro-odontoma transforming to ameloblastic fibrosarcoma [29].

Even though the occurrence of malignant transformation is rare [29] a strict follow up is recommended. An example of such a diligent control is mentioned in the study of Scheuer [23] where a patient was operated at age of 3.5 years and then followed up for more than 21.5 years without recurrence, however a 2 year follow up may suffice [13]. Follow up should be clinical and complimented with a radiological examination that would demonstrate the tumor adequately. Low resolution volume tomography at six months post operatively and then yearly for two years may be helpful in this regard and limit cumulative radiation in children.

4. Conclusion

In conclusion, the authors would like to contribute this unusual case of a young girl to further the knowledge regarding this odontogenic tumor, to recommend a conservative surgical management and a regular follow-up protocol for at least two years to rule out any evidence of recurrence or the rare malignant transformation [11,19,27].

Disclosure

None of the authors have any conflicts of interest that should be disclosed.

Conflict of interest statement

The authors state that there are no any financial, personal or organizational relationships that could inappropriately influence this publication.

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